であれば、比較的良好な知的予後が期待できる。後鼻孔狭窄による呼吸障害と頭蓋縫合早期癒合症が予後を左右する。

評価

1 新生児科、循環器科　睡眠時無呼吸の評価（ポリソムノグラム、酸素飽和度）睡眠時無呼吸の有無をポリソムノグラム、酸素飽和度により評価する。後鼻孔狭窄/閉鎖

3 整形外科・脳神経外科　頸椎の評価（側面頸部X線撮影、CT、

5 耳鼻科　聴力検査

6 歯科　咬合、歯、口腔内の評価（セファログラム、パントモ撮影、デンタル撮影）

泌尿器科　腎・泌尿器生殖器奇形を疑い腹部超音波

その他　尿検査（尿中ステロイドホルモンの定量）

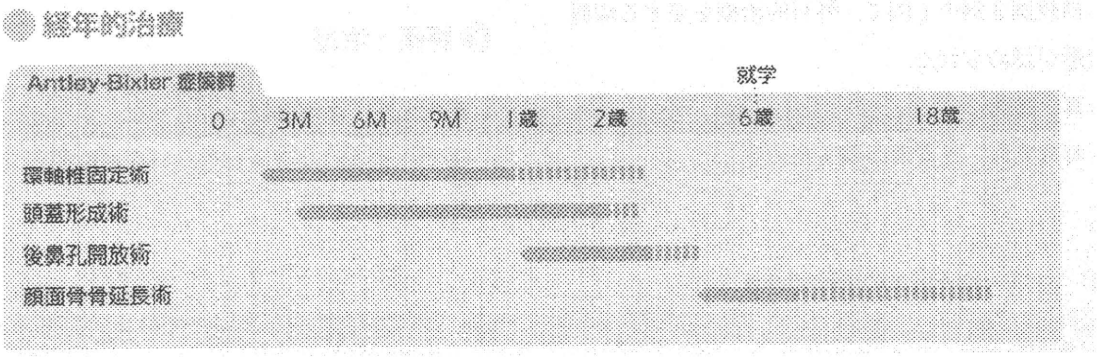
治療

1 環軸椎固定術

2 頭蓋形成術

3 歯科矯正治療

4 中顔面前方移動術（狭い上気道による睡眠時無呼吸の改善を含む）



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資料2　Syndromic Craniosynostosisに対する延長距離と方向を制御できる新しい顔面骨延長システム

要旨

現在でも、小児のsyndromic craniosynostosisに対して再現性良く延長量と延長方向を制御することは難しい。我々は、創外型および創内型骨延長装置の利点を併せ持った延長距離と方向を共に制御できる新しい顔面骨延長システムを開発した。その方法と短期結果について報告する。

方法

我々のシステムは従来の創外型骨延長装置と新しく開発した創内型延長装置の両者を使用する。

すなわち、術後における延長方向の制御は創外型で行い、延長距離の制御は角度可変型の創内型骨延長器で行う。本システムを2例のCrouzon syndromeに対して使用した。

結果

術中の合併症はなく、本システムにより、延長距離と方向を制御することができた。

結論

我々は、創外型延長装置と創内型延長装置の長所を生かしたシステムを開発し、延長距離と方向をともに制御することができた。多数回の再手術のために瘢痕が強い症例や.延長距離が20mm以上となる症例などは本システムの良い適応と考えられた。

眼窩周囲のプロファイルを合わせるために上顎がcounterclockwise lotationしたが、これらを解決するためには、Le Fort III型骨切り術とLe Fort IやII型骨切り術と組み合わせた術式が必要であると考えられた。

研究目的

顔面骨延長術は、下顎に初めて用いられて以来、craniofacial surgeryにおいて極めて重要なツールになっている。しかし、現在でも、小児のsyndromic craniosynostosisに対して再現性良く延長量と延長方向を制御することは難しい。この主な理由は、小児ではかなりのovercorrectionが必要であるだけでなく、inferior orbital rimとA点の延長すべき方向が異なっていることである。これに

対して現在では創外型と創内型の骨延長装置が使用されている。一般的に、創外型骨延長装置は延長方向の制御 control and adjustment of horizontal and vertical monements of the midfaceが可能だが、牽引する力 distraction forces for lengtheningが十分でないために延長距離に制限があり、顔面の十分なovercorrectionを行うことは難しい。一方、創内型骨延長装置は押し出す力が強いが、装着後の延長方向の制御が不可能である。我々は、これら両者の問題点を解決するために延長距離と方向を共に制御できる新しい顔面骨延長システムを開発した。その方法と短期結果について報告する。

方法

我々のシステムは創外型および創内型延長装置の両者を使用する。我々は、創外型として従来のハロー型骨延長装置 halo-type external distraction device(RED system, Martin LP, Jacksonville, FL,USA)を用いる。一方、創内型はハロー型骨延長装置と連動するために、新しく開発した3次元的に角度を変えられる骨延長装置を用いる。すなわち、本システムのconceptは、術後における延長方向の制御は創外型骨延長装置で行い、延長距離の制御は角度可変型の創内型骨延長装置で行う。

外科的手技

Syndromic Craniosynostosisに対する手術は、頭皮冠状切開によりLe Fort III型骨切術を行い、直径2mmのKirschner wireを頬骨-頬骨間に通した。このKirschner wireに角度可変型の創内型延長装置の前方部分を差し込んで固定した後に、後方部分を側頭骨にスクリューにて固定した。一方、創外型骨延長装置の牽引用Surgical wireは、左右計4本使用した。Surgical wireは、梨状口下縁に固定されたmini plateと頬骨-頬骨間のKirschner wireの2カ所に通され、創外型骨延長装置に取り付けられた。術後、中顔面骨は角度可変型の創内型延長装置によって両側の頬骨を貫通するKirschner wireを軸に前方へ押し出されることで延長距離が制御された。一方、創外型骨延長装置に取り付けられた左右計4本のSurgical wireにより延長方向が制御された(Fig.1-a,b)。延長直後に、創内型延長装置を残して創外型骨延長装置は抜去され、全てのSurgical wireは引き抜かれた。創内型骨延長装置は、約2ヶ月のconsolidation期間を経て抜去された。創内型骨延長装置の抜去は、まず頬骨のstab incisionから頬骨を貫通するKirschner wireを抜去した後に、5cmの側頭部切開を行い、装置を抜去するだけであった。頭皮冠状切開を用いないために、抜去術に要する時間は約20-30分であった。

症例

症例 1

6歳の男児　クルーゾン症候群。上顎と下顎の切歯の距離を示すOver jetは15mmであった。
新しいシステムを用いたLe Fort III型骨延長術が行われた。
手術5日後に1mm/dayのペースで延長が行われた。
延長終了の3か月後に全システムの抜去術が行われた。
側面頭部X線規格写真において延長距離は、オルビターレ（Or点）で28mm、A点で34mmであった。上顎はanticlockwise lotationしたが、the inferior orbital rim (orbitale)の位置はよかった。
症例2
8歳の女児。　クルーゾン症候群。上顎と下顎の切歯の距離を示すOver jetは15mmであった。
新しいシステムを用いたLe Fort III型骨延長術が行われた。
手術5日後に1mm/dayのペースで延長が行われた。
延長終了の3か月後に全システムの抜去術が行われた。
側面頭部X線規格写真において延長距離は、オルビターレ（Or点）で20mm、A点で26mmであった。上顎はanticlockwise lotationしたが、the inferior orbital rim (orbitale)の位置はよかった。

考察

RachmielらやStaffenbergらが、動物モデルにて顔面骨延長術が可能であることを示した。ヒトに対する顔面骨延長術は、下顎に初めて用いられて以来、craniofacial surgeryにおいて極めて重要なツールになっている。しかし、小児のsyndromic craniosynostosisに対して再現性良く延長量と延長方向を制御することは難しい。今回、我々は、創外型延長装置と創内型延長装置の長所を生かしたシステムを開発し、延長距離と方向をともに制御することができた。
創外型と創内型延長装置はそれぞれ利点と欠点を持っている。創外型の最大の利点は延長方向の制御が可能なことである。装置の取り付けと抜去は容易であるが、創外型の欠点は、創外型で平均26mmの延長距離を得たという優れた報告があるが、一般的には延長距離が限られることである。

一方、創内型では、延長方向の制御はできないが、延長距離の制御は可能であり、顔面骨を後方から押し出すことにより創外型より延長可能な距離は長く、小児のovercorrectionが可能である。創内に埋め込まれるために、装着中も比較的目立たずにすむことも利点の1つである。その反面、取り付けは創外型よりも煩雑で時間を要するだけでなく、左右のプレートのズレにより顔面の非対称性を生じる。一般的に、抜去する時は、頭皮冠状切開を行わなければならないことも欠点の一つである。固定されるプレート部分にbiodegradableな材料を使用したbiodegradable deviceはこれを解決

するための1つの手段である。また、小児ではzygomaticomaxillary sutureが脆弱であるために延長中に骨折することがある。創外型でレスキューした症例が報告されているが、従来の創内型ではレスキューすることができない。

本システムは、これらの問題点を解決するために開発された。本システムの利点は、1. 20-30mm以上の距離を延長することができるため、overcorrectionが可能である。結果として、手術回数を少なくすることができる。2. 3次元的に角度を変えることができる創内型骨延長装置は、たとえ左右対称に固定されていなくても、術後に方向の制御が可能である。先天的に左右非対称な症例にも応用可能である。3. 創内型の前方の固定はKirschner wireであるため、固定のためのthe body of zygomaの面積を必要としない。たとえ頬骨が低形成であっても取り付けることができる。4. consolidation期間を待たずに、延長終了直後に創外型骨延長装置を抜去することができる。5. 両側の頬骨を貫通するKirschner wireにより脆弱なzygomaticomaxillary sutureの骨折を予防することができる。6. 抜去は容易であり、頭皮冠状切開を必要としない。このように、本システムは多くの利点があり、ほぼ全てのSyndromic Craniosynostosisに対して適応があると思われる。特に良い適応は、多数回の再手術のために瘢痕が強い症例や.延長量が20mm以上になる症例などである。

今後の課題としては、小児でovercorrectionする時に、延長距離が伸びるほどthe inferior orbital rim (orbitale)やPoint Aの方向が異なるために、Cephalogram上の目標設定が難しくなることである。本症例でもthe inferior orbital rim (orbitale)を含めた眼窩周囲のプロファイルを合わせることを優先したために、上顎がcounterclockwise lotationし、Point Aは、理想よりも上方へ移動した。Apert syndromeに代表される顔面高が短い症例は、さらにthe orbital levelとocclusal levelsの延長距離と方向が異なることが予想される。これらを解決するためには、我々は、Le Fort IIIとLe Fort IおよびII骨切り術と組み合わせた術式やsplintとelastic tractionを組み合わせることによる両側の頬骨を貫通するKirschner wireを軸とする顔面骨の下方への回転を検討する必要があると考えている。

今後、我々は、これらの症例に対して注意深く中・長期的なfollow-upを行う必要がある。

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